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A Rare Case of Heterotopic Ossification in a Newborn: A Case Report

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ABSTRACT

Heterotopic ossification (HO) is the growth of bone in soft tissue, and can be broadly classified into neurogenic, genetic and traumatic causes. The pathophysiology of HO remains unknown. This disorder is extremely rare in infants and can mimic or coexist with thrombophlebitis, cellulitis or osteomyelitis. Most importantly, HO has to be differentiated from bone-forming tumours such as osteosarcoma and osteochondroma. We report a case of traumatic HO in a five-day-old newborn following intravenous cannulation of the right wrist and left ankle, with the latter complicated with osteomyelitis. We highlight the clinical and radiological features of HO and differential diagnoses of soft tissue ossification in early childhood.

Key Words:
Heterotopic ossification, newborn

INTRODUCTION

Heterotopic ossification (HO) is the growth of bone in soft tissue, where it does not normally exist [1]. HO was first described by Reidel in 1883, followed by Dejerne and Ceiller who observed HO in soldiers who sustained spinal cord injuries during the First World War [2]. Although the pathophysiology of heterotopic bone formation is not fully understood, the aetiology of HO is divided into neurogenic, genetic and traumatic causes [1,3]. The disorder is very rare in the paediatric population and almost unheard of in the newborn. Here we present a rare case of traumatic HO in a newborn following intravenous cannulation of the right wrist and left ankle, with the latter complicated with osteomyelitis. The patient's right wrist showed complete resolution following conservative therapy, as did the left ankle following drainage and six weeks of antibiotics.

CASE REPORT

A 3.28 kg. baby boy was admitted to the neonatal intensive care unit at birth because of neonatal hypoglycaemia and transient tachypnoeic of newborn, to rule out congenital pneumonia. The pregnancy was complicated by rupture of membranes for less than twelve hours prior to delivery. There was otherwise no maternal pyrexia or signs of chorioamnionitis. Delivery was by caesarean section at term because of maternal subfertility and gestational diabetes mellitus. Apgar scores were 7 and 9 at 1 and 5 minutes respectively.

The patient presented with swelling and erythema over the right wrist and left ankle at day five of life. These were previous sites of intravenous cannulation and venepuncture. Physical examination revealed a localized, firm and tender swelling over the radial aspect of the right wrist with surrounding warmth and erythema involving the distal third of the forearm. Similar findings were noted over the lateral aspect of the left ankle up to the lower third of the leg. The total white cell count was elevated at 28.3 (4-11 K/uL) with an absolute neutrophil count of 7.2 (2.0-7.5). C-reactive protein levels were less than 8mg/L. Blood culture and sensitivities did not grow any organisms from either site.

A preliminary diagnosis of thrombophlebitis of right wrist and left ankle was made and intravenous cloxacillin and C-penicillin were administered. Daily review showed signs of improvement. However five days later, the mother noticed whitish material discharging from the left ankle cannulation site. Plain radiography of the left ankle revealed hypertrophic ossifications in the lateral aspect of ankle joint. The visualized bones were normal with no periosteal reaction, bone sequestration or osteolytic areas to suggest osteomyelitis. Similar radiological findings were noted for the right wrist region. Two sets of blood culture did not grow any organisms.

A revised diagnosis of heterotrophic ossification of the right wrist and left ankle was made. Conservative management was employed with reassurance to the mother that spontaneous resolution is usually the case. The baby was discharged after having completed eight days of intravenous antibiotics, with a plan to complete syrup cloxacillin for another seven days.

The baby presented again at forty-six days of life with an erythematous swelling over the left ankle for two days, which eventually ruptured and discharged pus. His right wrist had improved. The white cell count was 11.9 (4-11

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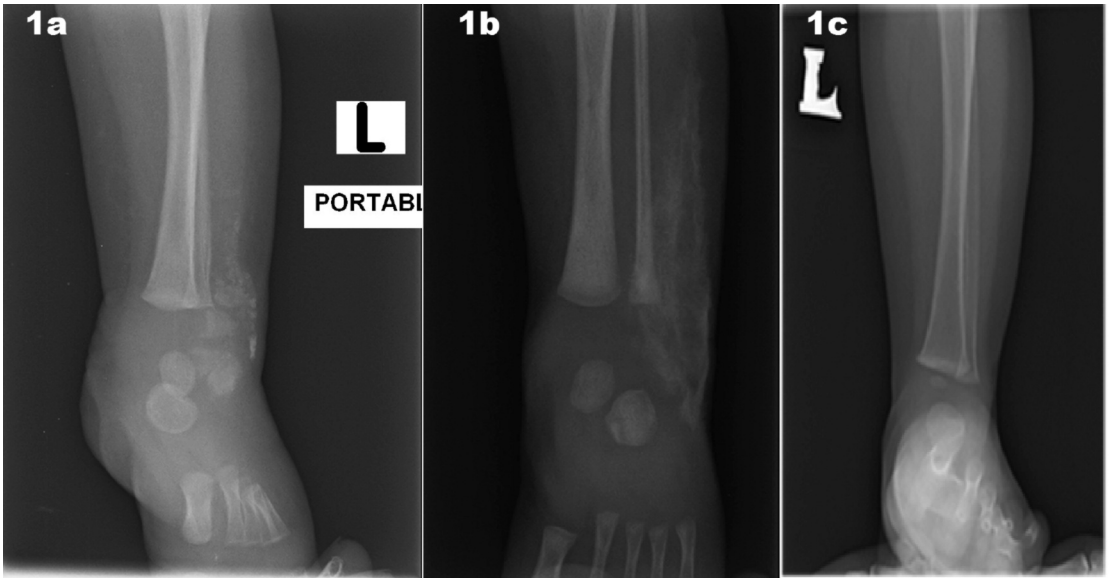

Fig. 1: Plan radiograph of left ankle showing initial amorphous calcified soft tissue lesion (1a), which evolved to a peripherally ossified mass surrounding a radiolucent, non-ossified centre one month later (1b) and complete dissolution at nine months (1c)

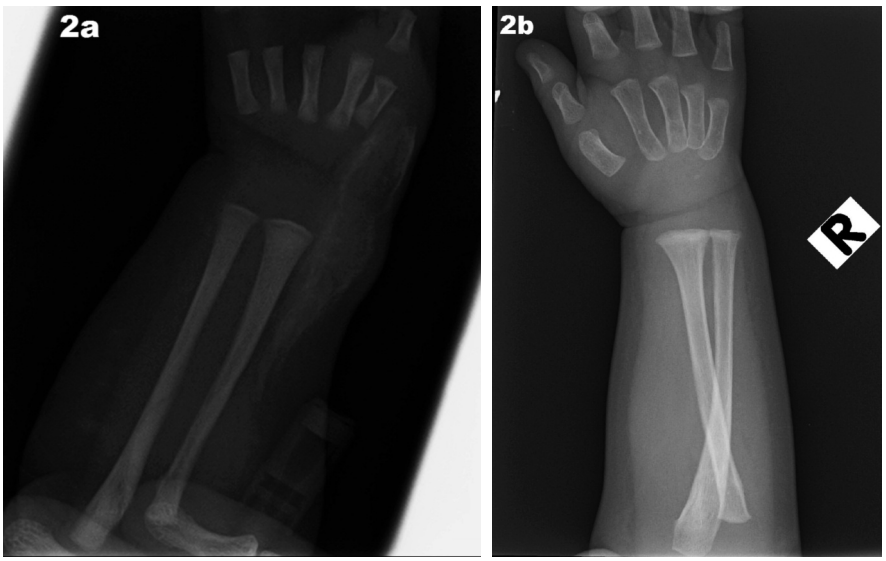

Fig. 2: Plain radiography of right wrist showing zoning phenomenon of peripheral maturation (2a) and regression of lesion with time (2b).

K/uL), absolute neutrophil count 2.1 (2-7.5) and C-reactive protein were elevated at 40 mg/dL. He was otherwise well, with no documented fever. Incision and drainage of the left ankle revealed unhealthy surrounding tissue with multiple areas of small calcified materials without pus or slough. Repeat plain radiography of the left ankle showed normal bone without periosteal reaction but more ossification in the surrounding tissue. In view of the fact that both pus and bone cultures resulted in moderate growth of *staphylococcus aureus* and *peptostreptococcus anaerobius*, the baby was treated for osteomyelitis of the left ankle with intravenous gentamicin and cloxacillin as per sensitivity results, for two weeks followed by oral cloxacillin for four weeks.

Subsequent clinic follow-up upon completion of six weeks of antibiotics showed complete resolution of the wound. The baby demonstrated some active range of movement in both affected joints. Review at nine months showed normal growth of both limbs with no limitation in range of motion. Repeat plain radiography showed complete dissolution of the right wrist and left ankle lesions.

DISCUSSION

Typical initial presentation of HO, an an acute inflammatory process, includes non-specific signs such as fever, swelling, erythema, warmth and reduced joint movement which can mimic or coexist with thrombophlebitis, cellulitis or osteomyelitis as seen in this case [1,4]. Diagnosis of HO in its initial stage is therefore difficult. We could have easily missed the diagnosis of HO if not for the whitish discharge from left ankle which called for further investigation. Conventional radiography may not show evidence of HO until 4 to 6 weeks after injury [1]. Three-phase bone scintigraphy is more sensitive for earlier detection of approximately 2.5 weeks after injury [1].

Diagnosis of HO should be reserved until it has been distinguished from other conditions which also cause soft tissue calcification. Ectopic calcifications can occur in hyperparathyroidism, sarcoidosis and dermatomyositis [5]. It is also commonly seen after subcutaneous fat necrosis following extravasation of calcium gluconate [5].

When present in the first decade of life, the existence of HO should raise suspicion of a rare, autosomal dominant disorder known as myositis ossificans progressive which is associated with progressive extraskeletal ossification and disfiguring manifestations. This chronic progressive disorder is readily recognized by associated microdactyly or adactyly of the first toe and thumb [1]. Our patient did not have any physical deformity. This petrifying condition demands careful management, and if suspected, bone biopsy or surgical excision should not be attempted as any trauma may aggravate the condition [1].

Although HO can sometimes be confused with osteosarcoma and osteochondroma, bone biopsy was not undertaken because we did not think it was necessary to subject the baby to an operative procedure since the clinical course and radiological findings were sufficient to distinguish HO from these conditions [1,4]. In HO, there should be a preceding inciting agent such as trauma (intravenous cannulation in this case), followed by spontaneous clinical resolution [4]. Radiographic features of HO include initial amorphous calcified soft tissue lesion which evolves to a radio-opaque, peripherally-ossified mass surrounding a radiolucent, non-ossified centre that shows regression with time [1,4]. This 'zoning' phenomenon of peripheral maturation is the most important diagnostic feature. HO does not involve the underlying bones and is extra-articular [1,4]. Ossification is seen between muscle planes and not intramuscularly [1]. The opposite is true for malignant bone tumours which often form dense central ossification [1]. Having said that, it would have been useful to send a sample for histopathological examination when the patient went for incision and drainage of left ankle. Unfortunately this was not done.

Treatment ranges from surgical excision, radiotherapy, or bisphosphonate to non-steroidal anti-inflammatory medications (NSAIDs) [1,3]. In the paediatric population, traumatic HO is usually managed conservatively with good prognosis for complete resolution [4]. In this case we believe the open wound from intravenous cannulation of the left ankle had probably introduced infection to the ectopic bone. Although the underlying bone was normal, we managed this cautiously as osteomyelitis with six weeks of antibiotics to minimize the possibility of spread of infection to the normal bone.

REFERENCES

1. Shehab D, Elgazzar AH, Collier BD. Heterotopic ossification. *J Nucl Med.* 2002 Mar; 43(3): 346-53.
2. Dejerne A, Ceillier A. Para-osteo-arthropathies des paraplegiques par lesion medullaire; etude clinique et radiographique. *Ann Med.* 1918; 5: 497.
3. Baird EO, Kang QK. Prophylaxis of heterotopic ossification - an updated review. *J Orthop Surg Res.* 2009; 2(4): 12.
4. Nissim L, Gilbertson-Dahdal. An unusual complication of an infiltrated intravenous catheter: Heterotopic ossification in a newborn. *J Radiol Case Rep.* 2008; 2(2): 13-5.
5. Dalinka MK, Melchior EL. Soft tissue calcifications in systemic disease. *Bull N Y Acad Med.* 1980; 56(6): 539-63.